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Shared Coronary Arteries and Coronary Venous Drainage in Thoracopagus Twins

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A pair of type B thoracopagus twins with complex cyanotic heart disease had shared coronary arteries and coronary venous drainage. Surgical separation was not attempted and the twins died at 10.5 months of age. Antemortem angiography demonstrated that Twin A's right coronary artery supplied Twin B's diaphragmatic and anterior ventricular myocardial free wall. A midline communication existed between each twin's right atrium at a common coronary sinus. The crossing coronary artery coursed alongside this connection and was visualized echocardiographically. At postmortem examination, the great cardiac vein of Twin A drained into the orifice of the common coronary sinus on Twin B's side of the midline.

In five of six previously reported cases, the children died at attempted separation shortly after ligation of the interatrial communication. This may have been because of occlusion of a coronary artery or acute obstruction of a coronary vein. Consideration of separation of type B thoracopagus twins requires anatomic delineation of the coronary arteries and veins.

Conjoined twins are rare, occurring approximately once in 50,000 to 80,000 live births (1,2). When joined at both the thorax and abdomen, they are called thoracopagus twins. Thoracopagus twins have union of the liver (100%), the heart (75%) and the gastrointestinal tract (50%) (3). Leachman et al. (4) identified three types of cardiac union: type A, common pericardial sac with completely separate hearts; type B, common pericardial sac with atrial connection only; and type C, common pericardial sac with both atrial and ventricular interconnections.

Modern surgical techniques allow separation of the musculoskeletal, hepatic and gastrointestinal communications. Type A cardiac union has posed few problems (3,5–9). Successful separation of type C cardiac union has not been reported and three attempts at separation of type B twins have resulted in five deaths. We present a case of type B thoracopagus twins, report the findings of shared coronary artery and vein systems and suggest that coronary vascular union may be the cause of previous failures at surgical separation. In our report, the terms right and left will be used only in relation to each twin such that right in Twin A is equivalent to left in Twin B. The terms anterior and posterior will be used in relation to the ventral and dorsal aspect of each twin, respectively.

Case Report

A pair of female conjoined twins was born at term in another hospital to a 29 year old mother. There was no family history of twinning, congenital heart disease or use of fertility drugs. Two months before delivery, abdominal ultrasonography identified twins, but the conjunction was not suspected. Cesarean section was performed because of active genital herpes. At birth, the twins weighed 5,500 g together. Apgar scores at 1 and 5 minutes were 5 and 7, respectively. Both babies were immediately given oxygen because of progressive central cyanosis and were transferred to Children's Hospital of San Diego for further evaluation.

Physical examination revealed a thoracopagus junction from the manubrium to a common umbilicus in which there were six vessels: two veins and four arteries. Mild central cyanosis during the breathing of 70% oxygen was present

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JACC Vol. 3, No. 4
April 1984:1019–25

in equal degree in both twins. No murmurs were audible in either twin. The pulses in all limbs were normal. Umbilical artery catheters were inserted in both twins and prostaglandin E_1 was administered to Twin B because of the clinical suspicion of ductal-dependent pulmonary blood flow.

Noninvasive Data

A lateral chest X-ray film demonstrated increased vascular markings in Twin A and decreased vessels in Twin B (Fig. 1). During breathing of 80% oxygen, arterial blood gases showed normal pH and partial pressure of carbon dioxide (P_{CO_2}) in both neonates; partial pressure of oxygen (P_{O_2}) values were 51 (Twin A) and 56 mm Hg (Twin B).

Leads I and aVF were recorded simultaneously from each twin followed by sequential electrocardiograms (Fig. 1). P-QRS complexes were synchronous, and the frontal plane QRS axes were superior in direction. Some of the QRS complexes were prolonged and slurred.

Two-dimensional echocardiograms. These were obtained from the standard short-axis, high long-axis and unconventional midchest and abdominal views in order to record intracardiac and great vessel anatomy. In Twin A, the great arteries were normally related (crossing) (10) and there was valvular and subvalvular pulmonary stenosis. Two ventricles and two atria, with a large atrial septal defect, were visualized. The ventricular apex pointed toward the right (dextrocardia). A small posterior ventricular cavity gave rise to the pulmonary artery while a larger anterior ventricular chamber gave rise to the aorta. Continuity was demonstrated between the aortic valve and the large anterior atrioventricular (AV) valve. The left-sided atrium received both a superior vena cava and pulmonary veins; the inferior vena cava and a second superior vena cava entered the right-sided atrium of Twin A.

In Twin B, the great arteries were malposed (parallel) (10) with the aorta larger than the pulmonary artery. The ventricular apex was located to the left (levocardia). A long, narrow subpulmonary chamber communicated with a large, functionally single ventricular chamber that received a single AV valve. Continuity between the single AV valve and either semilunar valve was absent. There was a large, functionally single atrium. The inferior and superior venae cavae entered a posterior atrial chamber, but the sites of pulmonary venous drainage could not be identified with certainty.

The anterior free walls of the ventricular masses of both twins appeared to touch in the region of the AV groove, but the main body and apex of each ventricular mass were separate. An intertwin atrial connection was visible across the midline (Fig. 2A). Injection of saline solution into a peripheral vein in the right arm of Twin A demonstrated opacification of two atrial chambers in Twin A; a few microbubbles crossed the midline to opacify the anterior atrium of Twin B. The reverse was demonstrated after injection into a peripheral vein in the right arm of Twin B. A large, continuous echolucent line appeared to cross the midline cephalad to the atrial connection (Fig. 2B).

Laboratory findings. Serum sodium, potassium, chloride, calcium, blood glucose and urea nitrogen levels were essentially identical in the twins, consistent with cross-cir-

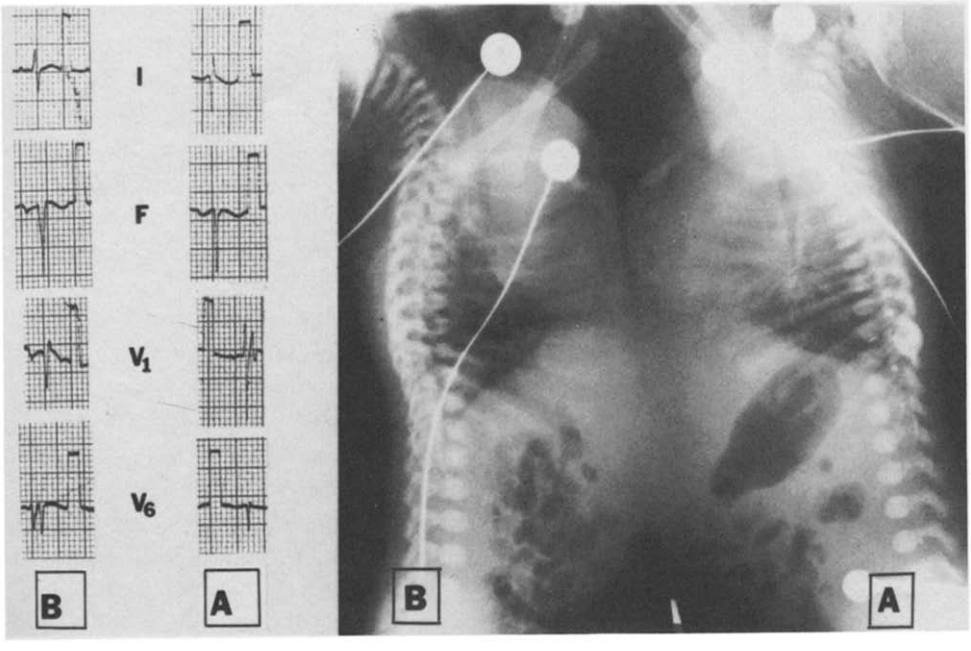

Figure 1. Admission electrocardiograms and chest X-ray film. [A] = Twin A; [B] = Twin B. **Left,** Admission electrocardiograms showing superior frontal plane QRS axis in both twins. The QRS complexes are slurred in leads V_6 (Twin B) and V_1 (Twin A). See text for discussion. **Right,** Admission lateral chest X-ray film showing thoracopagus union. Pulmonary vascular markings appear increased in Twin A and decreased in Twin B.

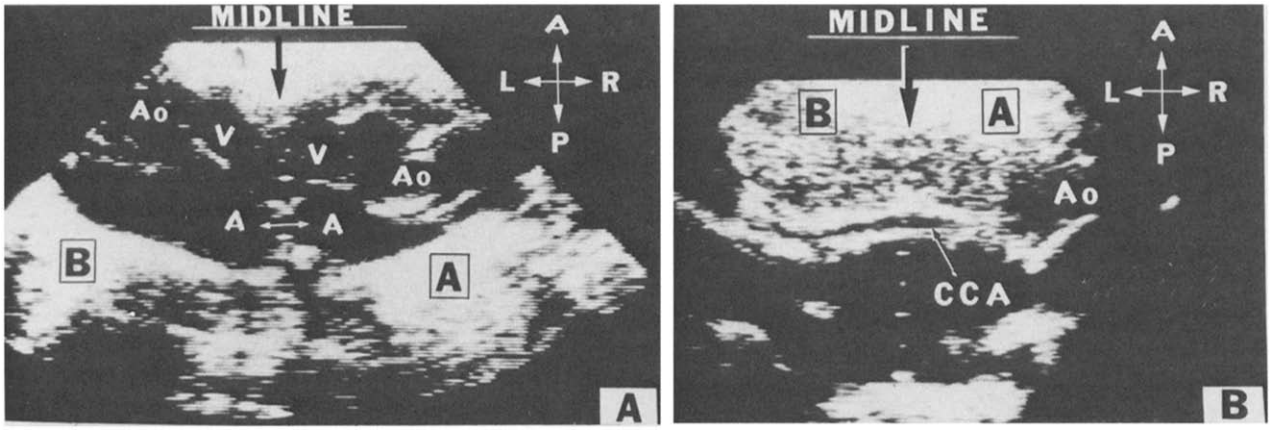

Figure 2. Two-dimensional echocardiograms. [A] = Twin A; [B] = Twin B. **Panel A,** The **bidirectional arrow** indicates the cross-midline intertwin interatrial (A↔A) communication. **Panel B,** Arising from the aorta of Twin A, a coronary artery crosses the midline and is (presumably) supplying Twin B. (See text and Fig. 4A and B.) A = anterior; Ao = aorta; CCA = crossing coronary artery; L = left; P = posterior; R = right; V = ventricle.

culation equilibration via the midline communication. After oral administration of activated charcoal to Twin A, the particles were found only in the feces of Twin A, confirming effective separation of the two alimentary tracts.

Cardiac Catheterization Studies

First study (36 hours). Simultaneous cardiac catheterization was performed in the twins at 36 hours and again at 4 months of age. Attempts to pass the venous catheter of Twin A across the midline into the anterior atrial chamber of Twin B, or vice versa, resulted in supraventricular tachycardia with slurred upstroke in the ipsilateral twin but synchronous, narrow QRS complexes in the contralateral twin, suggesting pre-excitation. Premature ventricular complexes introduced in one twin did not result in synchronous QRS complexes in the contralateral twin, indicating electrical separation of the ventricles. Systemic arterial pressure in Twin A was approximately 10 mm Hg higher than in Twin B (Table 1).

The anatomic findings noted noninvasively were confirmed by angiocardiography; pulmonary venous drainage was not well visualized.

Second study (4 months). At the second catheterization, systemic ventriculography in Twin A demonstrated a large "right" coronary artery that crossed the midline (Fig. 4A) and supplied the anterior myocardium of Twin B. Aortography in Twin B demonstrated a small posterior coronary artery which gave rise to several tiny branches that supplied the posterior and anterosuperior basilar myocardium of Twin B. Coronary vein drainage was not visualized. Surgery was not performed; the twins died at 10½ months of age at home.

Pathologic Anatomy

In each twin, the lungs contained two left and three right lobes. The gastrointestinal tracts were completely separate; there was a common liver, and a common pericardial sac enclosed both hearts. The combined heart-lung-liver weight was 597.5 g; projected weight for two normal female neonates is 864 g (11). The basic cardiac anatomy is shown in Figures 3 and 4. Only those aspects of anatomy not already determined by premortem studies will be discussed.

Right and left atria, systemic and pulmonary veins. Each child had two atria and two atrial appendages. In each child, a persistent left superior vena cava and all four pulmonary veins drained into the left atrium.

The midline intertwin atrial communication consisted of a tubular connection approximately 5 mm in diameter at its

Table 1. Cardiac Catheterization Data

	Oxygen Saturation (%)				Pressures (mm Hg)			
	Study 1*		Study 2†		Study 1*		Study 2†	
Site	Twin A	Twin B	Twin A	Twin B	Twin A	Twin B	Twin A	Twin B
RA	68		56		m = 11	m = 11	m = 9	m = 7
Systemic ventricle		84	59	64	74/15	62/17	96/12	76/7
Asc aorta			58	64		62/42	96/55	76/48
Desc aorta	88	84			74/50	67/42		

*FIO_2 = 0.8; †FIO_2 = 0.21. Asc = ascending; Desc = descending; m = mean; RA = right atrium.

JACC Vol. 3, No. 4
April 1984:1019–25

widest margin, joining the right atrium of each baby at a common coronary sinus. The orifice leading into the common coronary sinus on each side was guarded by a well-developed valve (Fig. 3B and C). The great cardiac vein of Twin A drained into the common coronary sinus on Twin B's side of the midline (Fig. 4C). The great cardiac vein of Twin B was not identified.

Ventricles, conotruncus and great arteries. These findings are presented in Figures 3 and 4.

Coronary arteries and veins. Twin A had three coronary ostia, two from the right coronary cusp (Fig. 4B) and one from the left (Fig. 4C). The more posterior right coronary ostium gave rise to a small short artery that divided into two branches supplying the proximal free wall and the interventricular septum of Twin A. The more anterior right coronary ostium gave rise to a single, very large coronary artery that coursed within the AV groove and continued across the midline within the adventitia of the atrial bridging tissue. After crossing the midline, this artery divided into four branches: two supplied the central anterior free wall of the single ventricle of Twin B; the third branch coursed within the AV groove to join the right coronary artery of Twin B at the aortic root (Fig. 4D); the fourth branch coursed over the apex of the ventricle of Twin B to join the left coronary artery of Twin B at the aortic root (Fig. 4B and E).

The left coronary artery of Twin A coursed within the left AV groove to supply the right and left ventricular outflow tracts and left atrium of Twin A (Fig. 4C). The great cardiac vein of Twin A was located in the right AV groove (Fig. 4C) and emptied into the common coronary sinus on Twin B's side of the midline.

Twin B had two coronary ostia: one from the right (Fig. 4D) and one from the left aortic sinus of Valsalva (Fig. 4E). The right and left coronary arteries of Twin B contained separate small branches that supplied the proximal, distal and left lateral free walls of the single ventricular chamber. The great cardiac vein of Twin B was not found. Gross communications with Twin A were as noted previously. Postmortem selective coronary angiography demonstrated coronary artery flow from the aortic root of Twin A to Twin B and vice versa.

Discussion

The approach to thoracopagus twins involves delineation of sites of intersystem communication (hepatic, gastrointestinal and cardiac) and the types of cardiac malformation. In those twins with a separate normal heart (type A of Leachman et al. [4]), surgical separation with survival of

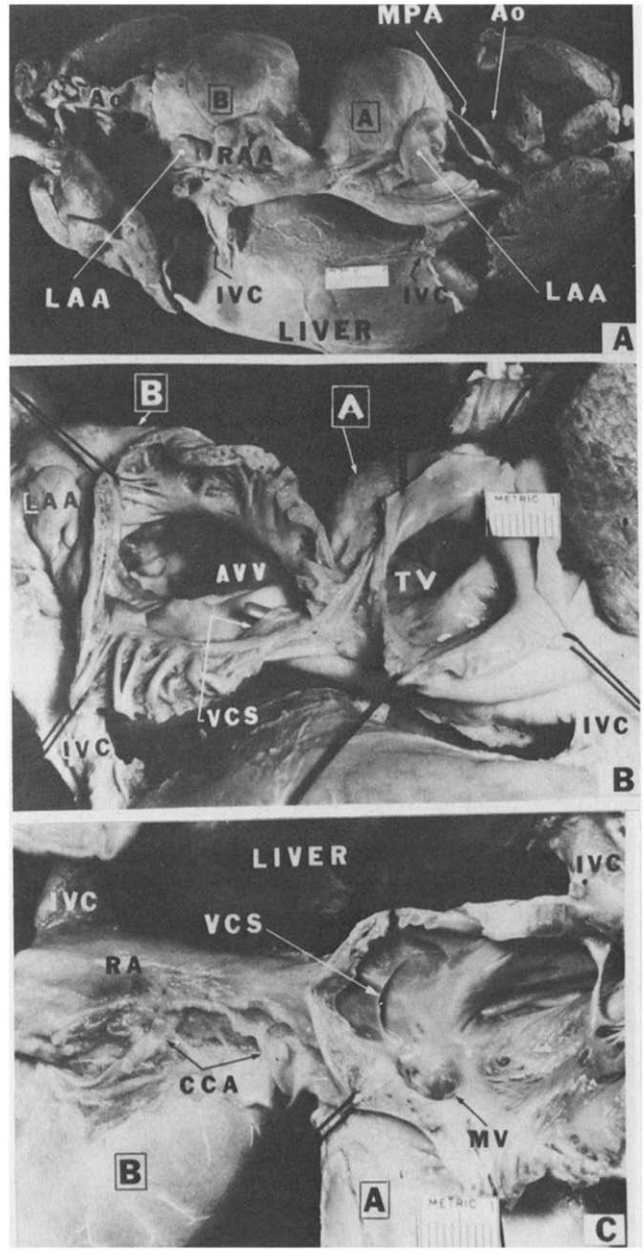

Figure 3. Pathologic anatomy. [A] = Twin A; [B] = Twin B; **Panel A,** Separate ventricular masses are seen with levocardia in Twin B and dextrocardia in Twin A. There is a common liver and a junction at the atrial level. Three atrial appendages (LAA and RAA) are seen on one side of the bicardiac mass. **Panel B,** The orientation is the same as in **Panel A;** the right atrium of Twin B and the left atrium of Twin A are opened. A probe tip traverses a common coronary sinus elevating the coronary sinus valve (VCS). **Panel C,** With the same orientation as in **Panel A,** the two hearts are rotated about the liver to display the opened right atrium (RA) of Twin A and the valve of the (common) coronary sinus (VCS). A stenotic mitral valve (MV) is seen and a crossing coronary artery (CCA) is identified in the adventitia crossing the midline. Ao = aorta; AVV = atrioventricular valve; IVC = inferior vena cava; MPA = main pulmonary artery; TV = tricuspid valve.

Table 2. Reported Type B Thoracopagus Twins

Reference (year)	Twin	No. of Atria	Location of Fusion	No. of AV Valves	No. of Ventricles	Great Vessels	PV Return	SV Return	Surgical Separation—Outcome
18 (1930)	A	2	Ra to LA of B	2	2	N	N	N	No; died
	B	2	LA to RA of A	CAVO	1	Truncus II or III	To Rt side (atrium)	To Lt side (atrium)	No; died
19 (1955)	A	?	LA to RA of B	?	?	N	?	R & LSVC; LSVC to LA	No; died
	B	?	RA to LA of A	?	?	N	?	R & LSVC; LSVC to LA	No; died
20 (1963)	A	2	RA to RA	2	2 + 2 VSD	N with PDA	N	N	No; died
	B	2	RA to RA	?	2 + 1 VSD	IAA-B	N	IVC, absent SVC	No; died
17 (1964)	A	2	RA to RA	2	2	N	N	N	No; died
	B	2	RA to RA	?	1	?	?	?	No; died
21 (1967)	A	1	?	1	1	D-malpos	?	?	No; died
	B	2 + ASD	?	2	2	N with PDA	N	?	No; died
13, 16 (1970)	A	2 + ASD	?	2	2 + VSD	N	?	?	No; died
	B	2 + ASD	?	CAVO	2 + VSD	DORV with IAA-A	?	Absent IVC	No; died
22 (1976)	A	2	?	?	?	?	?	?	No; died
	B	1	?	?	?	?	?	?	No; died
15 (1977)	A	2	CCS	2	1	D-malpos	?	?	No; died
	B	1	CCS	1	1	D-malpos + sub-PS	?	?	No; died
Present case (1983)	A	2	CCS	2	2	N with sub-PS	N	RSVC + IVC to RA; LSVC to LA	No; died
	B	2	CCS	1	1	L-malpos with sub-PS	N	LSVC to RA; RSVC to LA; IVC to RA	No; died
16, 13 (1970)	A	2	CCS	2	2	N	N	LSVC to CS; no RSVC	Yes; died
	B	2 + ASD	CCS	2	2 + VSD	N	N	N	Yes; died
15 (1977)	A	2	CCS	1	2; hypopl RV	Pulm Atr + PDA	TAPVR to CS	?	Yes; died
	B	2	CCS	2	2; hypopl LV	Aortic Atr + PDA	TAPVR to CS	?	Yes; died
14 (1979)	A	2	RA to RA	2	2	N with PDA	N	N	Yes; lived
	B	2	RA to RA	1	1	Pulm Atr + PDA	N	N	Yes; died

ASD = atrial septal defect; Atr = atresia; AV = atrioventricular; CAVO = common AV orifice; CCS = common coronary sinus; CS = coronary sinus; DORV = double-outlet right ventricle; hypopl = hypoplastic; IAA-A = interrupted aortic arch, type A; IAA-B = interrupted aortic arch, type B; IVC = inferior vena cava; LA = left atrium; LSVC = left superior vena cava; Lt = left; LV = left ventricle; malpos = malposition; N = normal; PDA = patent ductus arteriosus; PS = pulmonary stenosis; Pulm = pulmonary; PV = pulmonary venous; RA = right atrium; RSVC = right superior vena cava; Rt = right; RV = right ventricle; SV = systemic venous; TAPVR = total anomalous pulmonary venous return; VSD = ventricular septal defect; ? = not specified.

both twins is feasible. For type C twins (cardiac union at both atrial and ventricular levels), there is no currently available approach to separate and salvage both twins; to date, no survivor has been reported. Twins with type B union (atrial but not ventricular communication) usually have complex cyanotic heart disease (4,12); therefore, both the potential for separation and the appropriate treatment of the underlying heart defects in each child must be considered.

Diagnostic Considerations

Clinical features. Asymmetric pulses indicate separate ventricles; differences in blood studies may indicate the

absence of intercirculatory communications (either atrial or ventricular). Asynchronous P-QRS complexes suggest separate atria and ventricles (4,13). Slurred QRS complexes suggest separate ventricles but conjoined atria (4,13), as was true in our case. Synhorst et al. (14) reported the development of supraventricular tachycardia in one of a pair of type B thoracopagus twins after cardiac catheterization. Each of our twins developed supraventricular tachycardia during catheterization when the ipsilateral atrium was manipulated with the catheter. Such arrhythmias should be anticipated during noninvasive and invasive testing and surgical procedures.

Echocardiographic features. Two-dimensional echocardiography provides extensive information about both the specific cardiac malformations and the presence of intercirculatory communications. In addition to standard echocardiographic views, unconventional views and contrast echocardiography are necessary for a complete study. In an attempt to demonstrate intercirculatory communications, venous contrast injections should be performed in both arms of each child because there may be a persistent right or left superior vena cava draining into an atrium that does not communicate across the midline.

Role of invasive studies. Cardiac catheterization may be indicated in all cases of thoracopagus twins. Even when type A cardiac union is suspected, the lack of normal echocardiographic leads, chest X-ray views and echocardiographic planes makes it difficult to be sure of normal cardiac anatomy.

Importance of Coronary Anatomy

Coronary artery anatomy. Our case experience indicates that in addition to atrial, ventricular and great vessel anatomy, cineangiocardiography must be directed specifically at coronary artery anatomy and flow. Failure to identify the coronary artery supply in type B hearts may result in unexpected death in either or both twins. Three attempts to separate type B twins have been reported (Table 2). Each pair had right atrium to right atrium communication; in two of the three (15,16), atrial connection at a common coronary sinus was confirmed at autopsy. In the remaining case (14), the location of the right atrial union was not specified. Coro-

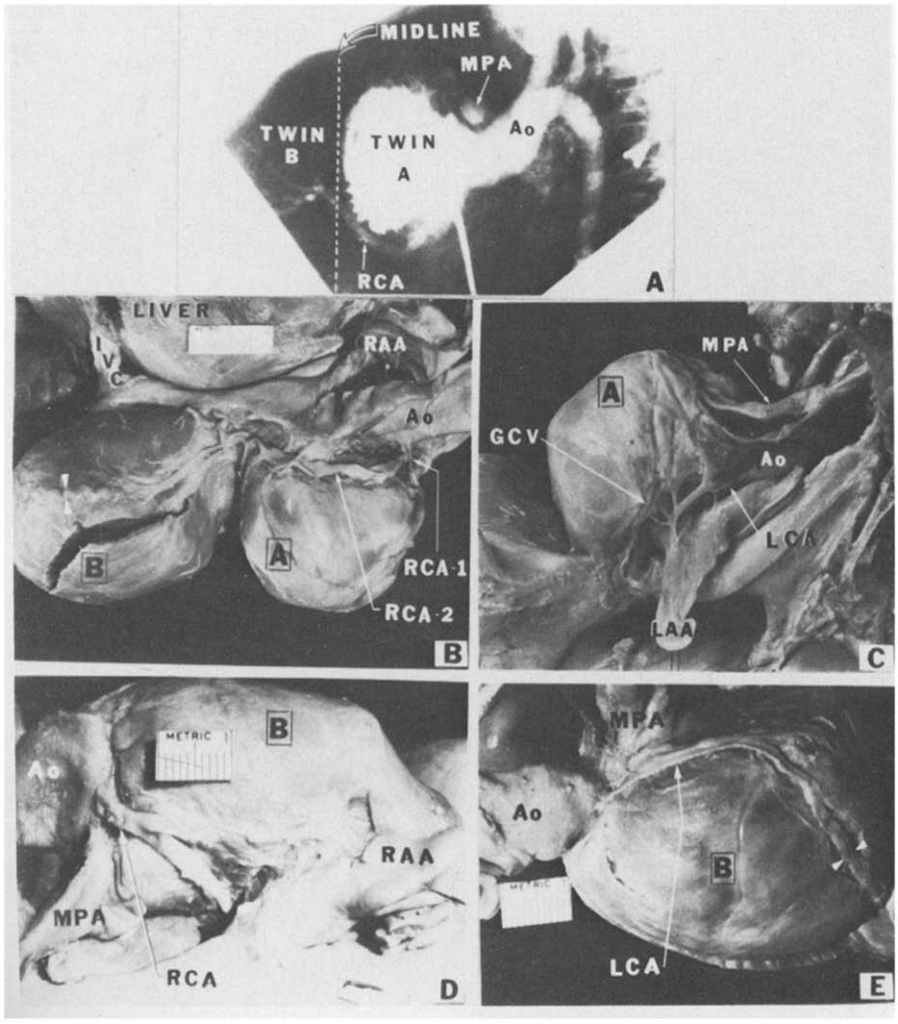

Figure 4. Coronary artery anatomy. **Panel A,** Right ventricular angiogram fills aorta (Ao) and a right coronary artery (RCA) which crosses the midline to supply the myocardium of Twin B. **Panel B,** Two right coronary arteries arise from the right sinus of Valsalva of Twin A. The larger, more anterior one (RCA-2) is the vessel seen in **Panel A** crossing the midline; the smaller right coronary artery (RCA-1) supplies the anterior myocardium of Twin A. The **arrowheads** identify a coronary vessel on the heart of Twin B which joins the RCA-2 of Twin A with the left coronary artery (LCA) of Twin B (see Panel E). **Panel C,** The left atrial appendage of Twin A has been retracted to expose the left coronary artery (LCA) and the great cardiac vein (GVC). **Panel D,** The right coronary artery (RCA) of Twin B has two branches. **Panel E, Arrowheads** indicate the coronary vessel connecting the left coronary artery (LCA) of Twin B with the right coronary artery of Twin A. IVC = inferior vena cava; MPA = main pulmonary artery; RAA = right atrial appendage.

nary artery and vein anatomy was not described in any of the twins. Of the six potential surviving children, there was only one long-term survivor (14); this child's twin died with ventricular fibrillation 7 days after operation. Death was attributed to renal, liver and cardiac failure. In each of the other four deaths, one twin died immediately after ligation of the atrial communication and the other died within hours after surgery. These four deaths could be attributed to coronary artery ligation or creation of coronary vein obstruction. In the absence of contradictory autopsy findings, it is possible that the death of Twin B as reported by Synhorst et al. (14) could also have been due to similar causes.

Type B twins with shared coronary artery supply may be separable if bilateral cineangiocardiography demonstrates a balanced coronary artery blood flow from each aortic root. In our case, although pathologic study and postmortem selective coronary arteriography revealed that each aortic root communicated with the other by way of a bridging coronary artery, in vivo angiography demonstrated coronary artery flow from Twin A to Twin B but not from Twin B to Twin A.

Coronary venous drainage. This is an additional complicating factor in separation of type B thoracopagus twins. Twelve pairs of type B twins have been reported including the present case (Table 2); a common coronary sinus was present in 4, probable in 3 and possible in 3 of the 12 pairs. Therefore, when considering separation of type B twins, viability will depend on preservation of both coronary artery flow and coronary venous drainage. Simple ligation of atrial bridging tissue may occlude a critical crossing coronary artery and obstruct coronary venous flow. Although coronary artery anatomy and flow can usually be delineated by angiography, identification of coronary venous drainage remains a formidable task. If not seen during the venous phase of selective coronary arteriography, coronary venous drainage may not be completely visualized even at surgery judging from our postmortem dissection experience.

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